

Case Report

Bilateral inverted vertebral arteries (V3 segment) in a case of congenital atlantoaxial dislocation: Distinct entity or a lateral variant of persistent first intersegmental artery?

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Abstract

Background: Anomalous vertebral arteries (VAs), commonly involving the persistent first intersegmental artery (FIA), are often seen with congenital atlantoaxial dislocations (AAD). Here we describe an unusual variant consisting of bilateral VAs with normal loops but passing below the C1 (inverted VA) arch, distinctly different from the FIA.

Case Description: A 9-year-old boy presented with a spastic quadriparesis. Preoperative radiographic studies showed an irreducible AAD with an occipitalized CO-C1 and C2-3 fusion. Although both VAs exhibited proximal and distal loops like normal VA, the distal loops did not pass through the C1 transverse foramina and coursed inferior to the C1 arch instead. With this critical preoperative information, both VAs could be better safeguarded during dissection of the C1-2 facets.

Conclusion: In the case presented, although the course of the inverted VAs is similar, the norm, they coursed inferior to both C1 arches. Careful evaluation of the preoperative radiological studies allowed for careful dissection of the inverted VA (horizontal loop) while opening the C1-2 joint for subsequent alignment (e.g. reduction) and bony fusion. This information also facilitates safer insertion of lateral mass screws (e.g. choosing the appropriate C1 screw length to gain adequate bony purchase without compromising anomalous VA).

Key Words: Bilateral, injury prevention, operative steps, persistent first intersegmental artery, vertebral artery anomaly

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INTRODUCTION

A major anomaly of the vertebral artery (VA), involving the persistent first intersegmental artery (FIA), is often encountered with congenital atlantoaxial dislocations (AAD).^[3,4,6,9] Here we present an unusual anomaly involving both VA wherein the proximal and distal loops were normal, but coursed inferior to the C1 (inverted VA) arch.

CASE REPORT

A 9-year-old boy presented with a progressive spastic quadriparesis following trivial trauma. X-ray studies showed an occipitalized CO-C1 and congenital C2-3 fusion; both contributed to an irreducible AAD resulting in cervico-medullary junction compression. Of interest, the C1 sagittal inferior facet angles measured 138°. The computed tomography (CT) angiogram clearly defined

the anomalous course of the V3 segments of both VA; the VA exited from the C2 transverse foramen, took a lateral

course toward the C1 transverse foramen, but instead of traversing the C1 foramen, turned inferiorly and coursed

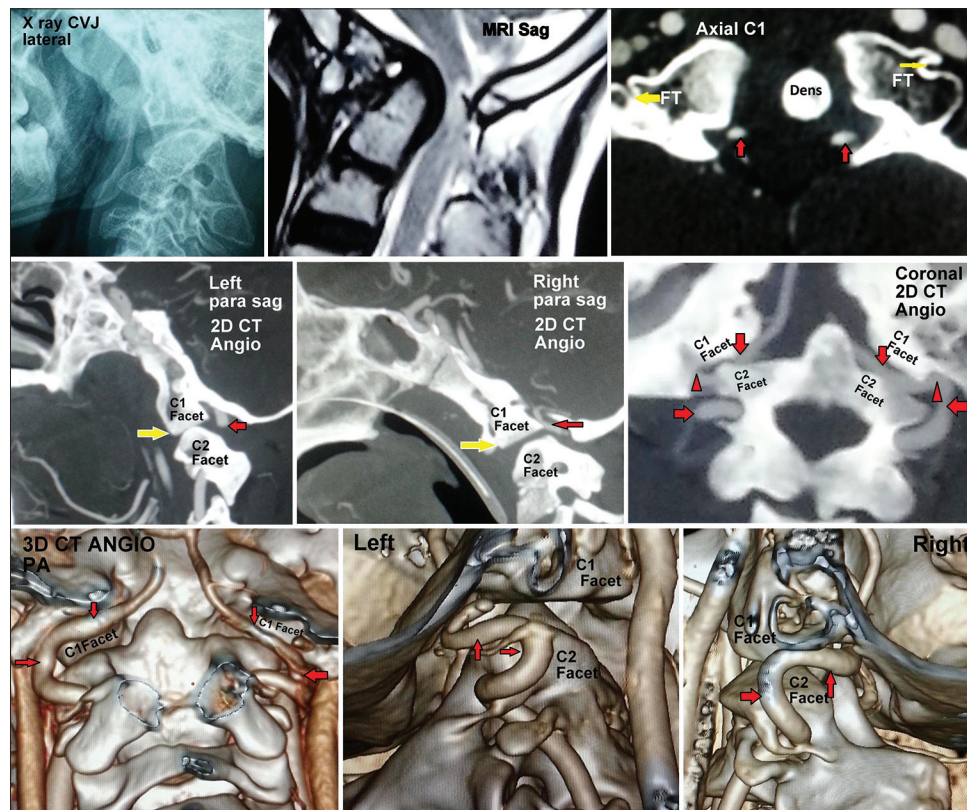


Figure 1: Preoperative images – Top row: X-ray and MRI of CVJ showing AAD. Adjacent to it is axial CT angio image showing C1 transverse foramina without vertebral artery (yellow arrows). VAs are seen medially (red arrows). Middle row: 2D CT angio images in parasagittal and coronal plane passing through C1-C2 joint showing acute C1 inferior sagittal facet angle (yellow arrow) and bilateral inverted VA (red arrows). Bottom row: 3D CT angio showing bilateral inverted VA (red arrows) in relation to C1-C2 facet joint. The course of VA is akin to normal except that it passes inferior to transverse foramina and C1 facet

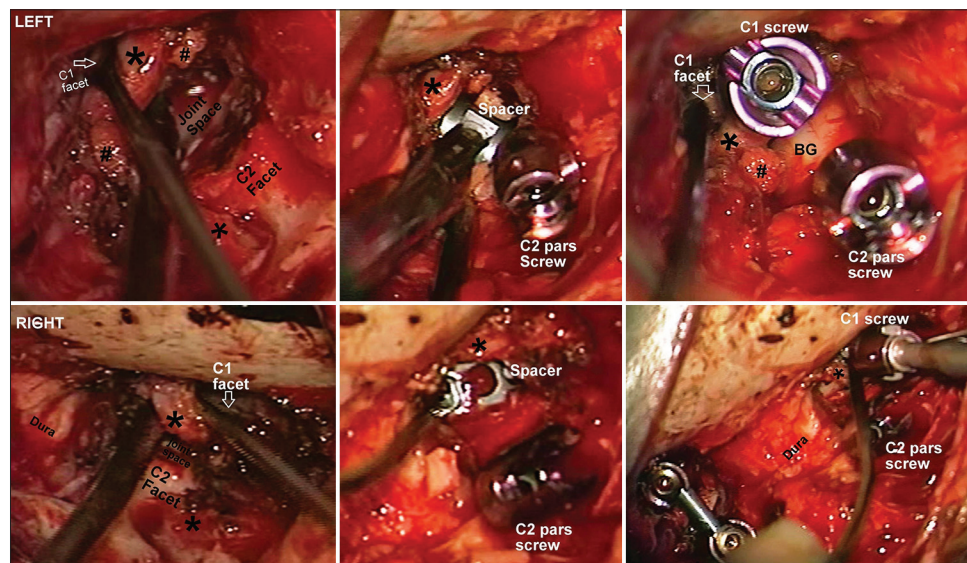


Figure 2: Intraoperative images showing Bilateral anomalous VA's (*) and its horizontal course posterior to inferior C1 facet. The course can be traced from C2 transverse foramen and then ventral to the C2 root ganglion (# cut C2 root), forming a horizontal redundant loop posterior to the inferior C1 facets. Images show sequential exposure of bilateral C1-2 joints and placement of spacers with bone graft (BG). Note the gentle inferior retraction of the anomalous horizontal VA loop to expose the inferior C1 facet and placement of C1 screws leaving enough space to avoid its compression by the screw head

medially beneath the C1 posterior arch before piercing the dura [Figure 1]. Therefore, both VA lay posterior to the inferior facet of C1 (3D CT; Figure 1).

Surgical decompression/fusion

As the patient was incompletely reduced with traction, surgical intervention was warranted. The C1-2 joints were approached posteriorly, and both C2 nerve root ganglia were cut to expose the anomalous VAs just anterior to it. In order to avoid inadvertent injury to the VAs, the loops crossing the C1 facets were carefully exposed; the posterior wedge of the C2 and anterior wedge of the C1 facets were drilled making the inferior facet angle flat in order to reduce the AAD. Spacers and bone grafts were placed in the C1-2

joint spaces. Next, the VAs were gently retracted inferiorly to insert the C1 lateral mass screws, which were sufficiently tightened to leave enough space between the screw head and the facet surface for the VA; the postoperative CT also showed adequate reduction [Figure 2 and 3]. Postoperatively, the child did well, becoming ambulatory, and 4 months later, the CT exhibited good bony fusion.

DISCUSSION

The VAs usually course postero-laterally after exiting the C2 transverse foramen, forming a short proximal loop that then enters the transverse foramen of the atlas. It then courses obliquely on the dorsum of the posterior arch of atlas forming the distal loop before penetrating the dura [Figure 4].^[2,8]

VA anomalies

The anatomical variations involving the VA between C1 and C2 have been well described.^[3,4,6,9] One variant, the FIA, usually courses medially and cranially after exiting the C2 transverse foramen to pierce the dura; it lacks the proximal and distal loops of a normal VA [Figure 4]. In the “inverted” VA variant presented, both VA had normal proximal and distal loops, but the proximal loop reached just short of the C1 transverse foramen [Figure 3]. A unilateral ectatic VA with similar anomalous course has been described in the past.^[4]

Embryology of the VA

Embryologically, the VA is formed by vertical channels interconnecting the cervical intersegmental arteries. During the course of time, these intersegmental arteries disappear and vertical connections persist.^[1] The vertebrobasilar junction and intracranial VA is formed from multiple channels.^[5]

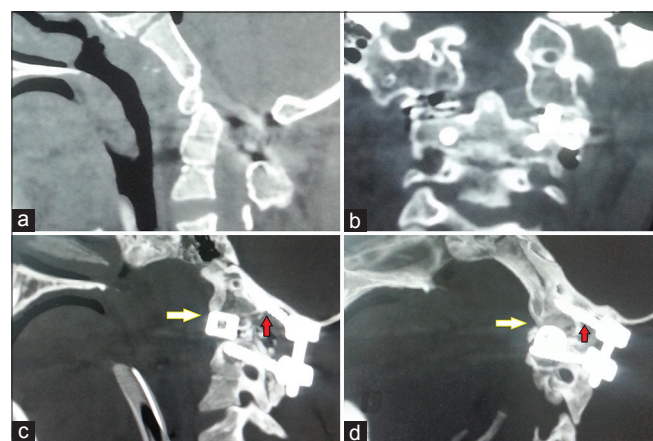


Figure 3: (a and b) Midsagittal and coronal CT images showing complete reduction of Atlanto-axial dislocation. (c and d) parasagittal CT images passing through right and left C1 and C2 facets with screws and spacers. Note the flat drilled surface of C1 facets (white arrows) as compared with preoperative. Also note the adequate space between the posterior surface of inferior C1 facet and the screw head, enough to house the anomalous VA loop without compressing it

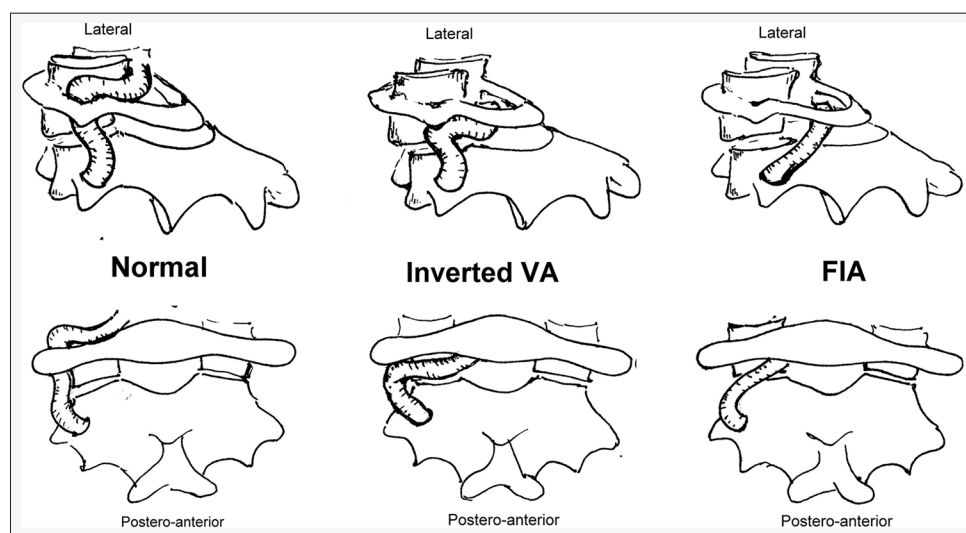


Figure 4: Comparison of normal VA, upside down VA and persistent first intersegmental artery (FIA). Note the presence of short proximal and a horizontal distal loop both in normal and upside down VA. These loops are absent in FIA, which, directly crosses medially after exiting C2 transverse foramina. The upside down VA reaches just short of the C1 transverse foramina and the horizontal loop lies below the C1 unlike the normal VA

Anomalous VA

The anomalies of VA are commonly seen distal to the C2 transverse foramen and are comprised of persistent channels.^[5] If the FIA persists alone, it leads to FIA. If the connecting channel persists with the FIA, it leads to fenestration. The connecting channels may exist both cranial and caudal to the first sclerotome. If both the cranial channels and the FIA become blocked for some reason, the caudal channels may persist giving rise to an 'inverted VA'. Nevertheless, an alternative hypothesis is that lateral traction on the FIA gives rise to the 'inverted VA'.

Anomalous VA pose a surgical challenge

VA anomalies pose a surgical challenge as these anomalous vessels may be injured during drilling of facet surfaces, placement of spacers, and the insertion of lateral mass screws.^[6] The FIA obliquely/medially crosses the joint space, and therefore, makes it difficult to open the joint space and drilling the facet surface, but provides good exposure for C1 facet screw insertion. Alternatively, the "inverted" VA needs to be dissected all along its horizontal course and retracted inferiorly so to facilitate exposure of the posterior surface of C1 facet to allow for safer screw insertion. Furthermore, the screw length should be sufficient to attain good purchase, but should not compress the VA.

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